Urology Case Reports 6 (2016) 1–3

Contents lists available at ScienceDirect

Urology Case Reports

journal homepage: www.elsevier.com/locate/eucr

Oncology

Primary Mixed Glandular-endocrine Tumor of the Male Proximal Urethra: A Case Report

Fernando N. Facio [a,*], Luis C. Spessoto [a], Germano J. Ferraz de Arruda [a], Jeronimo F. Arruda [b], Jose G. Ferraz de Arruda [a], Maria Fernanda W. Facio [c]

[a] FAMERP, Brazil
[b] UNILAGO, Brazil
[c] FACERES, Brazil

ARTICLE INFO

Article history:
Received 12 December 2015
Accepted 16 December 2015

Keywords:
Urethra
Mixed tumor
Malignant
Neoplasms

ABSTRACT

Primary urethral carcinoma is much more common in women than in men due to its association with urethritis. A 65-year-old man presented with a 10-month history of penile induration, obstructive voiding symptoms and hematuria. Urethrocystoscopy showed a solitary mass in the proximal urethra and no bladder involvement. It was performed penectomy without bladder neck excision and regional staging lymphadenectomy of the obturator lymph nodes. Pathological diagnosis revealed adenosquamous cell carcinoma with squamous and glandular components. We described a case of primary mixed glandular-endocrine tumor of the male proximal urethra consisting of adenosquamous cell carcinoma with squamous and neuroendocrine components.

Introduction

Primary urethral carcinoma is much more common in women than in men due to its association with urethritis, being the ratio of female to male predominance reported as 4:1.[1] Among the different tumors arising in the male urethra, squamous cell cancer is the most frequently occurring histological type (80%), the remaining being mainly transitional cell carcinoma (15%) and adenocarcinoma or undifferentiated carcinomas (5%).[1] Other, rarer cell types include lymphoma, sarcoma, paragangliomas, melanoma, and neuroendocrine carcinoma.[1]

We present a case report related to primary mixed glandular-endocrine tumor of the male proximal urethra.

Case report

A 65-year-old smoker man presented with a 10-month history of penile induration, obstructive voiding symptoms and hematuria. He had no inguinal lymphadenopathy. Investigation was conducted by the Urology Service of Hospital de Base, São José do Rio Preto, and started in May 2008. Urethrocystoscopy revealed a solitary mass of 2.5 × 3.5 cm in the proximal urethra and no bladder involvement. On magnetic resonance imaging (MRI) of the abdomen and pelvis the lesion extended into the corpus spongiosum and corpora cavernosa. We performed penectomy (Fig. 1) without bladder neck excision and regional staging lymphadenectomy of the obturator lymph nodes. Pathological diagnosis was made based on histology and immunohistochemistry. They revealed adenosquamous cell carcinoma (Fig. 2A) with squamous and glandular components arising in the urethral mucosa among a heavy inflammatory infiltrate. Surrounding tissues did not show lymphangiosis carcinomatosa but the tumor had positive resection margin. Neoplastic cells were immunoreactive for neuron-specific enolase (NSE), cytokeratin 20, p63 protein (Fig. 2B) and chromogranin A (CgA) (Fig. 2C) that were compatible with neuroendocrine differentiation. Further examination demonstrated a tumor of the lung that was initially thought to be a metastasis, so palliative chemotherapy (cisplatin plus irinotecam) was initiated. The patient underwent lobectomy that resulted in a pT2pN0M0 squamous cell carcinoma (SCC). Chemotherapy was discontinued because his immune status was impaired and unfortunately he died in April 2010 from septic complications related to pneumonia.

* Corresponding author.
E-mail address: fnfacio@yahoo.com.br (F.N. Facio).

http://dx.doi.org/10.1016/j.eucr.2015.12.007

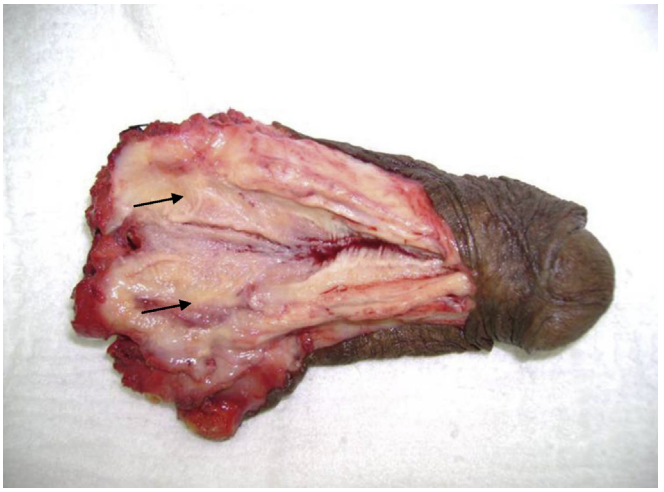

Figure 1. Penectomy specimen with 3.5 cm tumor in the proximal urethra.

Discussion

We described a case of primary mixed (composite) glandular-endocrine tumor of the male proximal urethra consisting of adenosquamous cell carcinoma with squamous and neuroendocrine components. Specific markers that may be used to establish neuroendocrine differentiation comprise NSE, CD56, CgA and synaptophysin, being the two latter recommended due to their relative sensitivity and specificity.[2] Composite tumors arise through multidirectional differentiation of a single neoplasm.[3] We believe that the urethral neoplasm in our case was the primary tumor and pluripotent stem cells of the urethral epithelium capable of neuroendocrine differentiation were the most likely source of origin. Although endocrine-paracrine cells are known to exist in the urethroprostatic region, they are considered postmitotic and incapable of neoplastic transformation.[4]

Computerized tomography and ultrasonography have a limited ability to detect penile or urethral tumor invasion of the corpus spongiosum or corpora cavernosa. On the other hand, MRI not only provides superior tissue contrast, but also permits imaging in any plane and to identify distinctly local lesion extension.[2] In our case, staging resulted in local cancer disease, so only penectomy was performed. For advanced cancer disease, the literature is unable to draw specific treatment recommendations based upon the current studies. However, chemotherapy selection can be determined from case reports that demonstrated cervical glassy cell carcinoma, a poorly differentiated form of adenosquamous carcinoma, to be sensitive to carboplatin and paclitaxel.[5]

The pulmonary SCC finding, once considered a metastasis, is probably another primary tumor because it consists of a single and unilateral mass without glandular differentiation in a patient with a long-time smoking addiction. The mucicarmim, an histochemistry staining that is used to highlight any kind of mucin produced by normal or neoplastic cells, was also negative in this tumor. Moreover, there was no local or lymph node metastases by the urethral neoplasm, thus the possibility of the pulmonary lesion, with a so distinctive morphology, being a distant metastasis of that is virtually impossible.

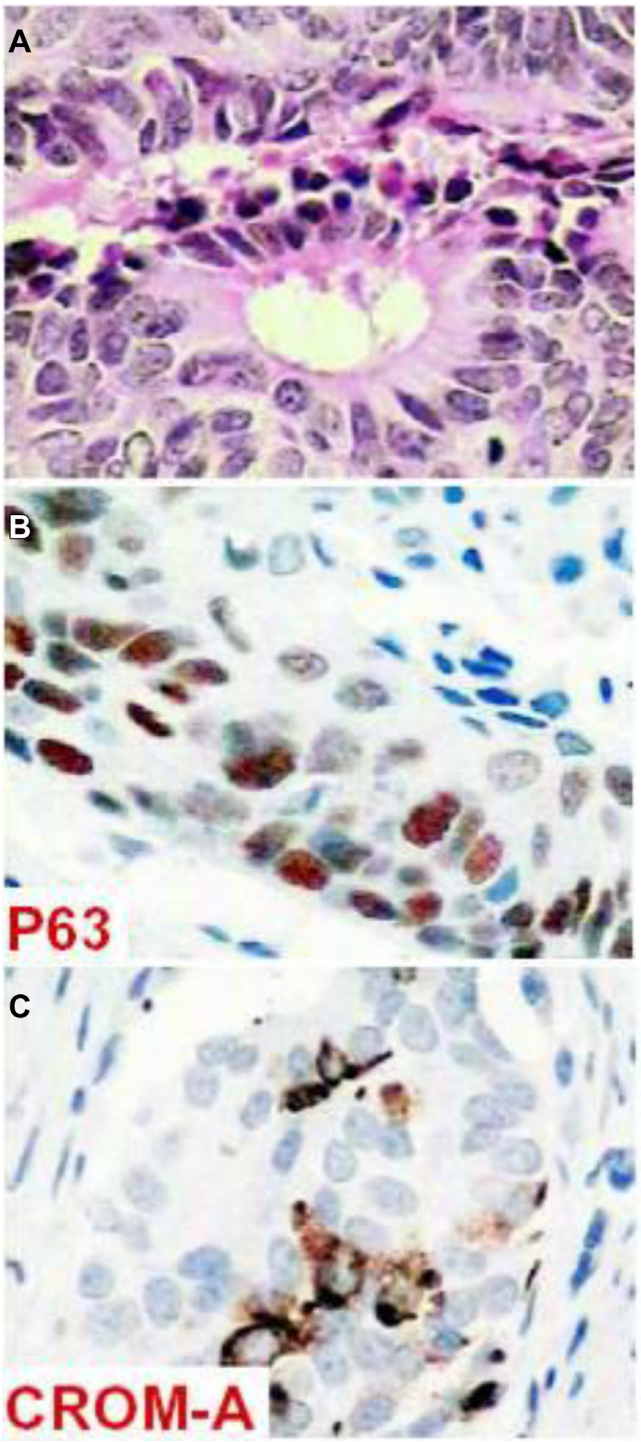

Figure 2. Carcinoma (A); neoplastic cells for p63 protein (B) and CgA (C).

We are the first to report adenosquamous cell carcinoma with squamous and neuroendocrine components arising in the urethral mucosa. It also represents the unique case of a concurrent primary SCC of the lung which can represent a link of predisposing factors, such as smoking. This neoplasm should be considered in the differential diagnosis of focal urologic malignancies.

Conflict of interest

There is no conflict of interest.

References

1. Yoo KH, Kim GY, Kim TG, et al. Primary small cell neuroendocrine carcinoma of the female urethra. *Pathol Int*. 2009;59:601–603.
2. Washington MK, Tang LH, Berlin J, et al. Protocol for the examination of specimens from patients with neuroendocrine tumors (carcinoid tumors) of the colon and rectum. *Arch Pathol Lab Med*. 2010;134:176–180.
3. Eble JN, Sauter G, Epstein JI, Sesterhenn IA. *World Health Organization Classification of Tumors. Pathology and Genetics of Tumours of the Urinary System and Male Genital Organs*. Lyon: IARC Press; 2004.
4. Vadmal MS, Steckel J, Teichberg S, Hajdu SI. Primary neuroendocrine carcinoma of the penile urethra. *J Urol*. 1997;157:956–957.
5. Weissbart SJ, Bianco Jr FJ, Sotelo T, et al. Glassy cell carcinoma of the urethra. *Urology*. 2009;73:60.